Case Report

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Capillary Hemangiomas of the Temporalis Muscle. Technical Report and Review of the Literature

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ABSTRACT

Introduction: Hemangiomas are benign vascular tumors defined by an atypical proliferation of blood vessels. These lesions may develop in any vascularized tissue, including the skin, subcutaneous layers, muscles, and bones. Intramuscular hemangiomas account for less than 1% of all hemangiomas and predominantly occur in the musculature of the trunk and extremities. However, involvement of the temporalis muscle is exceedingly uncommon.

Material and Methods: This study presents two newly identified cases of capillary hemangiomas affecting the temporalis muscle, diagnosed using CT, MRI, and arteriography

Results: The surgery was perform with preoperative selective embolization of the donor vessels.

The hemangiomas were resected without any complications, the patients are free of disease five and nine years after surgery

Discussion: Of 41 cases reported in the literature of hemangiomas of the temporalis muscle, only 37 are well documented. Cavernous hemangioma is the most common type with 30 cases; 4 are capillary and 2 are mixed type. Surgical excision was performed in 34 cases and 3 cases underwent clinical and radiological follow-up. Preoperative embolization was performed only in 1 case of capillary hemangioma. Clinical and radiological findings, as well as treatment options are discussed.

Keywords: Capillary; Hemangioma; Temporalis Muscle; Arteriography; Embolization

Introduction

Hemangiomas are benign vascular neoplasms, accounting for approximately 7% of all benign tumors. Hemangiomas are most commonly found in the skin and subcutaneous tissue. Intramuscular hemangiomas (IMH) account for less than 1% of all hemangiomas and are predominantly located in the muscles of the tunk and extremities [1]. IMH in the head and neck region make up less than 15% of these cases, with the masseter muscle being the most frequently involved site [2-4]. However, IMH of the temporalis muscle are exceedingly rare, with only 41 cases documented in the literature to date (Table 1) [5-40]. Three categories of hemangiomas have been identified based on the type of vessels involved: capillary, cavernous and mixed [41]. Cavernous type is the most frequent of the IMH of the temporalis muscle reported.

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Table 1: Reported instances of temporalis muscle hemangiomas.

Cavernous Hemangioma					
Case	Author/Year	Sex	Years	Treatment	Embolization
1	Joehl, et al. [6]	F	59	Surgery	No
2	Knox, et al. [7]	M	19	Surgery	No
3	Murakami, et al. [9]	M	51	Surgery	No
4	Hughes, et al. [10]	M	28	Surgery	No
5	Couloigner, et al. [11]	F	41	Surgery	No
6	Tada, et al. [12]	F	14	Surgery	No
7	Cappabianca, et al. [13]	F	13	Surgery	No
8	Lopez-Cedrun, et al. [14]	M	41	Surgery	No
9	Itosaka, et al. [15]	F	12	Surgery	No
10	Shpitzer, et al. 1997	F	29	Surgery	No
11	Benateau, et al. [16]	F	61	Surgery	Yes
12	Sharma, et al. [17]	F	5	Surgery	No
13	Sherman, et al. [18]	M	1	Surgery	No
14	To, et al. [19]	F	54	Surgery	No
15	Heckl, et al. [20]	F	55	Follow-up	No
16	Bui-Mansfield, et al. [21]	M	44	Surgery	No
17	Sakr, et al. [23]	M	44	Surgery	No
18	Calisaneller, et al. [25]	M	37	Surgery	No
19	Bucci, et al. [26]	M	38	Surgery	No
20	Kim, [27]	M	24	Surgery	No
21	Gadhia, et al. [28]	F	57	Follow-up	No
22	Eryilmaz, et al. [29]	M	34	Surgery	No
23	Turel, et al. [31]	F	61	Surgery	No
24	Cui, et al. [32]	M	62	Surgery	No
25	Kishimoto, et al. [34]	M	43	Surgery	No
26	Jbali, et al. [35]	F	42	Surgery	No
27	Motazedian, et al. [36]	M	64	Surgery	No
28	Watanabe, et al. [37]	F	69	Surgery	No
29	Sayad, et al. [38]	F	37	Surgery	No
30	Alqahtani, et al. [39]	F	45	Surgery	No
31	Jafarian, et al. [40]	F	51	Surgery	No
Capillary Hemangioma					
Case	Author/Year	Sex	Years	Treatment	Embolization
32	Sharma, et al. [8]	M	21	Surgery	No
33	Benateau, et al. [16]	F	61	Surgery	yes
35	Sharma, et al. [17]	M	27	Surgery	No
34	Arora, et al. [33]	M	5	Surgery	No

Mixed Hemangioma					
Case	Author/Year	Sex	Years	Treatment	Embolization
35	Top, et al. [22]	M	46	Surgery	No
36	Kim, et al. [30]	F	46	Surgery	No
Unknown					
Case	Author/Year	Sex	Years	Treatment	Embolization
37-40	Scott, [5]	N/A	N/A	N/A	N/A
41	Uraloglu, et al. 2005	F	41	Follow up	No

Material and Methods

Case 1

A 31-year-old man was referred to our Department for management of a left temporal swelling of at least 18 months duration which had been gradually increasing in size causing facial asymmetry. No history of trauma in the left temporal region was reported. Examination revealed a soft, not well circumscribed lesion about 2'5 cm of diameter in the left temporalis fossa. The mass seemed to move in relation with temporalis muscle movements. The overlying skin was normal. No bruits or pulsation were detected, and no pain associated. There was no cranial nerve deficit or cervical lymphadenopathy. The imaging diagnose began with a computed tomography (CT) which revealed a solid, soft tissue mass in the left temporal region that extends to the infratemporal fossa and towards the pterygopalatine fossa, without signs of bone or skin infiltration, and an uniform enhancement after contrast administration. Magnetic resonance imaging (MRI) was performed to achieve a correct diagnosis, and showed a soft tissue lesion in close relation to the temporalis muscle, isointense on T1 weighted sequences and a high signal after paramagnetic contrast administration (Figure 1).

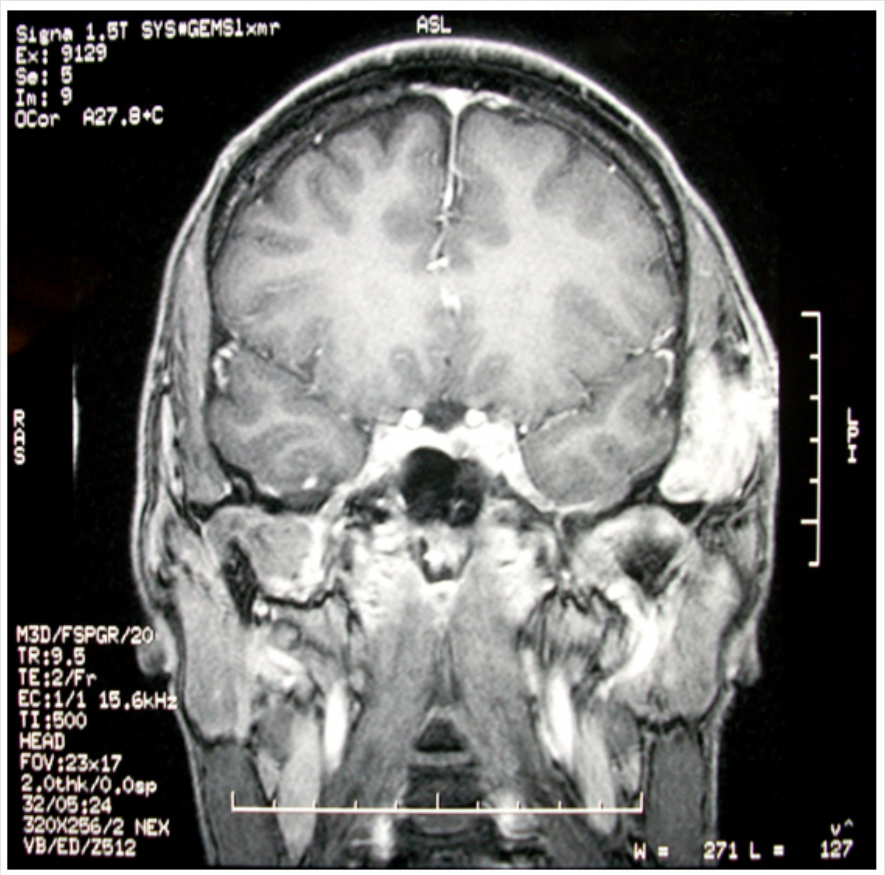

Figure 1: Magnetic resonance image showing a soft tissue lesion in the left temporalis area.

Due to aesthetic concerns and continuous growth, surgical excision was recommended. Before the surgery, a subselective arteriography was performed. In the pre-embolization images, a hypervascularized mass depending on branches of the left internal maxillary artery was identified (Figure 2). The tumor was embolized using polyvinyl alcohol microparticles, and liquid coils in the left internal maxillary artery's pedicle, showing significant decrease of the lesion in the post-embolization images (Figure 3). One day following embolization, the patient underwent surgical removal of the hemangioma using a hemicoronal approach with preauricular and submandibular incisions. External carotid artery was temporally clamped due to the possible intraoperative bleeding. The left temporalis muscle within the angiomatous mass was then totally excised to the coronoid process, after removal of the zygomatic arch and ligation of the left maxillary artery at the liquid coil's level. The zygomatic arch was then replaced, and the defect filled with a temporal polyethylene implant. On the histological examination, the tumor was composed of a large number of interconnecting endothelial-lined vascular spaces in skeletal muscle, consistent with an intramuscular haemangioma of the capillary type. The patient had an uncomplicated postoperative recovery, achieving favorable functional and cosmetic outcomes. Nine years after the surgery, there are no signs of recurrence

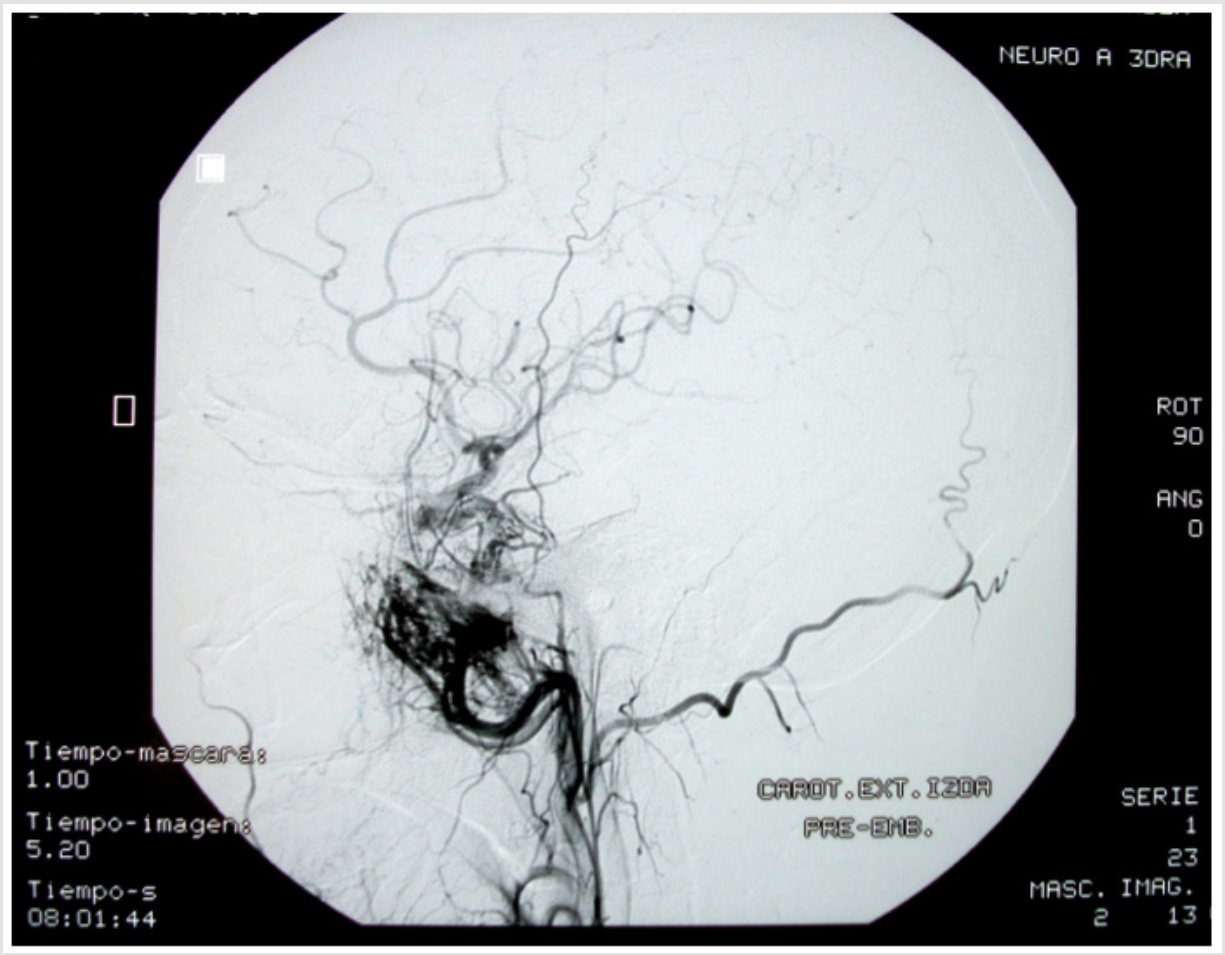

Figure 2: Subselective arteriography of the left external carotid artery. Pre-embolization image showing a hypervascularized mass depending on branches of the left internal maxillary artery.

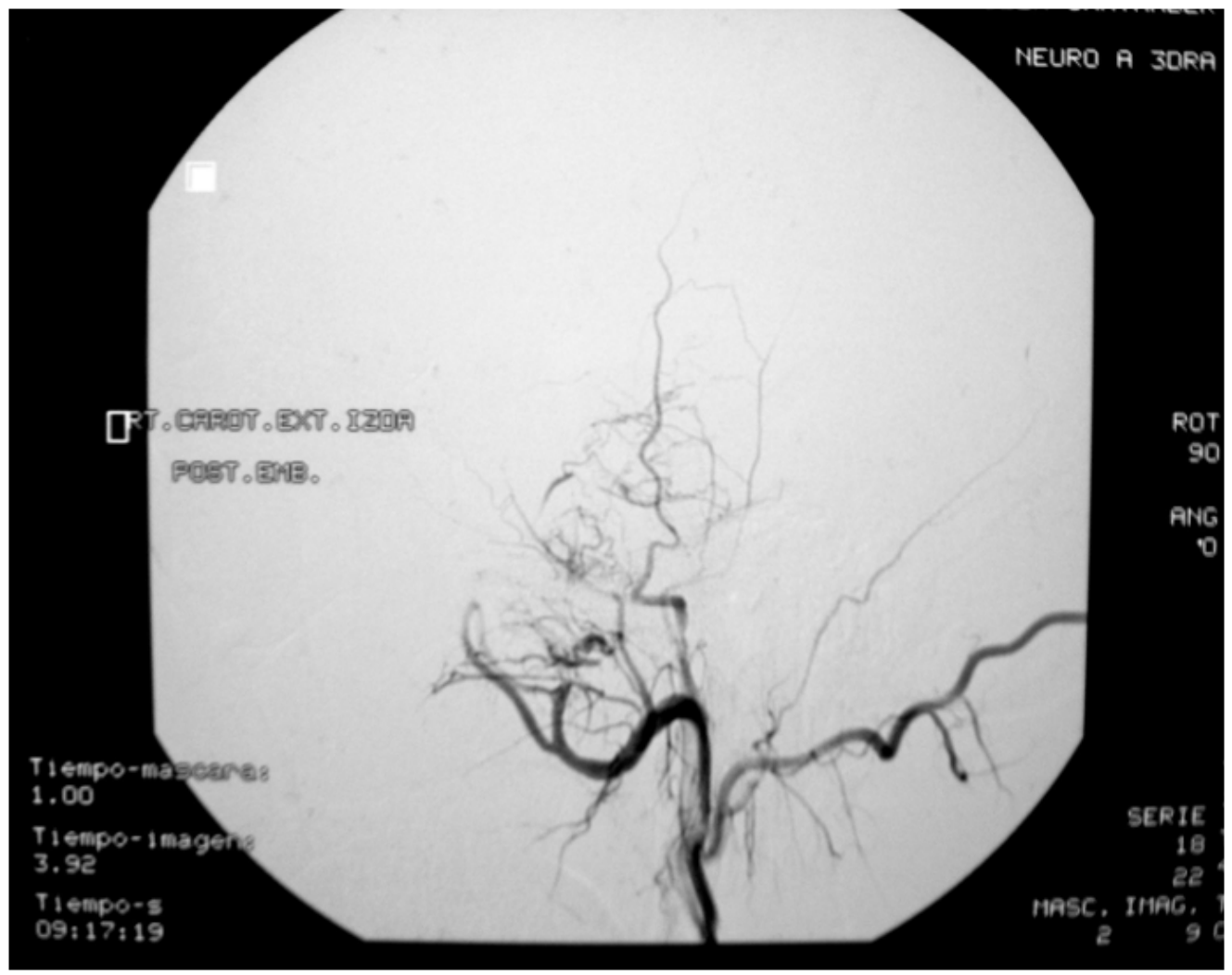

Figure 3: Post-embolization image with significant decreasing of the lesion.

Case 2

A 35-year-old woman with a 3-month history of a slow growing mass in the left temporal fossa. There was no pain, but the patient felt itches occasionally during night. On the physical examination there was a soft and tender swelling in the left temporal region, with a slight pulsation palpable. No bruits were detected, and the overlying skin was also normal. Imaging workup included ultrasonography (US) and MRI. US showed a 6x17mm solid, ovoid mass in the left temporal region with intense vasculature. On MRI, the tumor appeared as a mass within the left temporalis muscle, isointense on T1 images and markedly hyperintense on T2 sequences, with vigorous enhancement after gadolinium administration (Figure 4). The preliminary diagnosis was of hemangioma of the temporalis muscle, and surgical excision under general anesthesia after embolization was planned. Subselective arteriography showed two vascular masses depending on the left deep temporal artery, one with a high-flow arterio-venous fistula (Figure 5). After embolization of the lesion and the fistula with biologic glue and microparticles, and occlusion with coils of the deep temporal, superficial temporal and internal maxillary arteries, the hemangioma virtually disappeared (Figure 6). Surgical approach to the temporalis muscle was achieved through a hemicoronal incision 24 hours after the embolization. The muscle within the hemangioma was totally excised after deep temporal branches localization and zygomatic arch osteotomy, which was later replaced (Figure 7). The defect was filled with a temporal polyethylene implant. The biopsy results confirmed a diagnosis of capillary hemangioma in the temporalis muscle. The patient was asymptomatic after the procedure, with good functional and cosmetics results. The 5-years-follow-up didn't show signs of recurrence.

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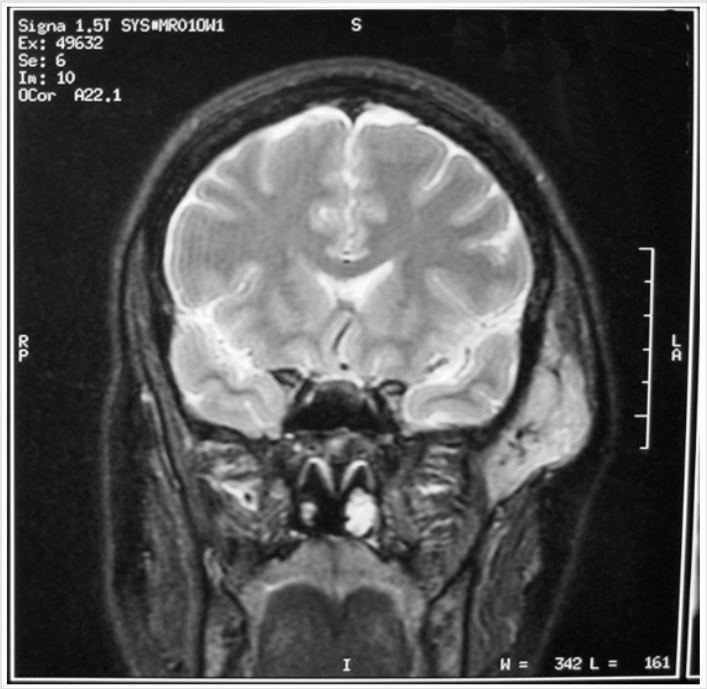

Figure 4: MRI showing a mass within the left temporalis muscle.

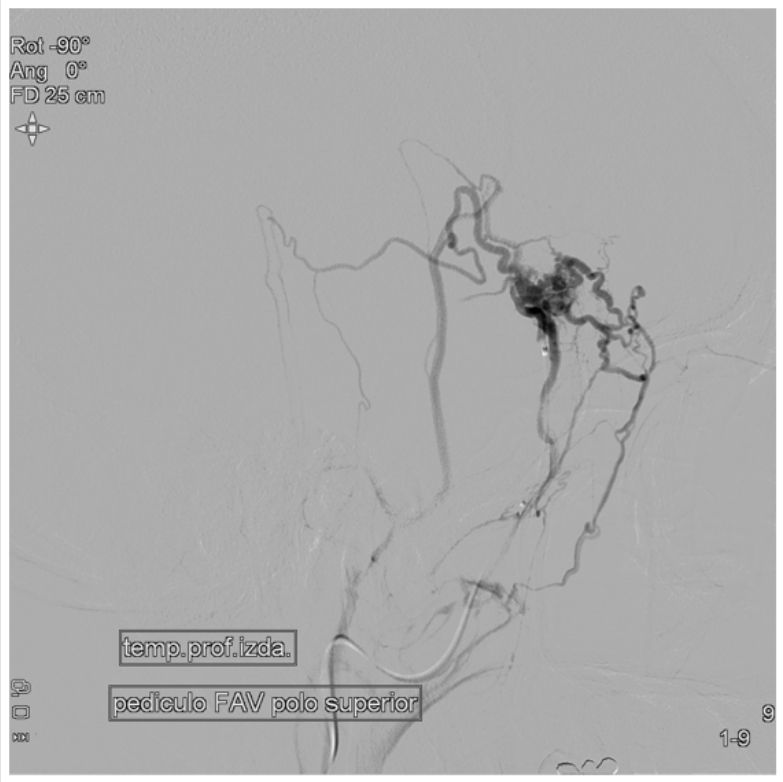

Figure 5: Subselective arteriography of the left external carotid artery.

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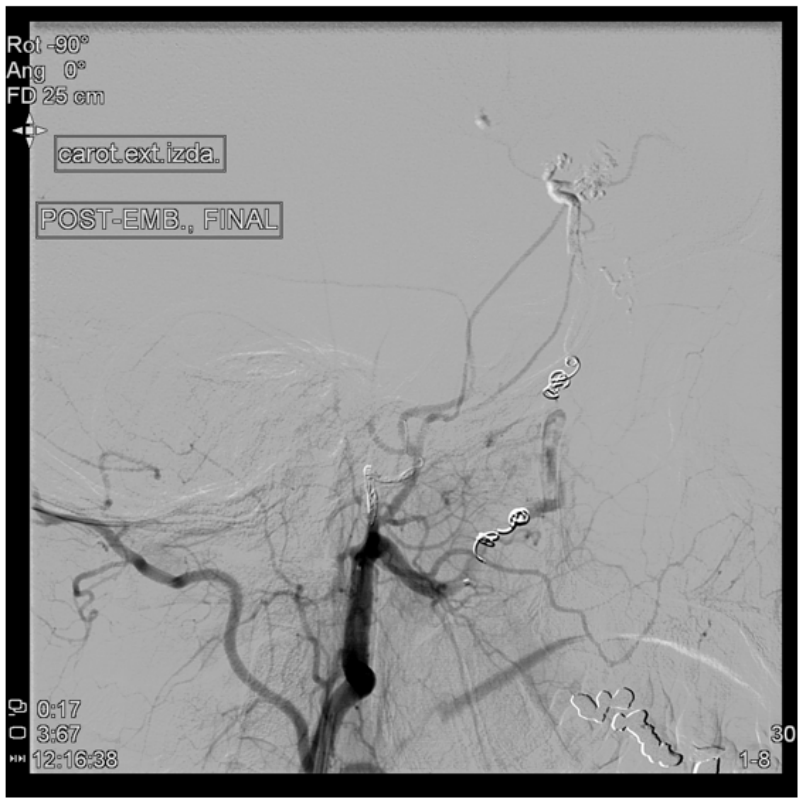

Figure 6: Post-embolization of the high-flow arterio-venous fistula. Final post-embolization image.

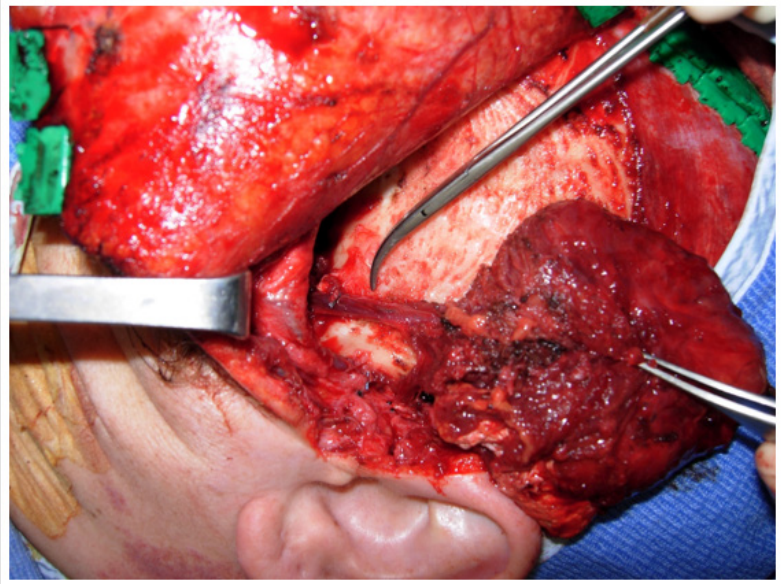

Figure 7: Left temporalis muscle excision after deep temporal branches localization.

Discussion

Intramuscular hemangiomas (IMH) are uncommon tumors, accounting for about 0.8% of all hemangiomas. Approximately 14% of IMH cases are found in the muscles of the head and neck[1]. The most commonly affected muscles in this region include the masseter (36%), trapezius (24%), periorbital muscles (12%), and sternocleidomastoid (10%) [2-4]. However, the temporalis muscle is an extremely uncommon site for these tumors, with only 41 cases documented in the literature to date. Of these, only 37 cases have been thoroughly described in detail [5-40]. IMH were initially described by Liston in 1843, [42] and later classified by Allen and Enzinger in 1972 according to vessel size. Three types are recognized: small vessel or capillary type (diameter < 1.40 μm), large vessel or cavernous type (diameter >140 μm); and mixed type, which includes both [41]. In the head and neck region, small-vessel hemangiomas represent approximately 30% of IMH, while large-vessel and mixed types account for 19% and 5%, respectively. However, within the temporal muscle, cavernous hemangioma is reported as the most common type according to the literature. Interestingly, both of our cases were diagnosed as capillary-type hemangiomas.

Approximately 50% of intramuscular hemangiomas (IMH) develop during the first decade of life, with 94% occurring before the age of 30 [43]. There is no distinct gender preference, as the literature reports 18 males and 19 females, with ages ranging from 1 to 69 years (mean age: 38.4 years)."The etiology of these lesions remains unknown. They are believed to result from abnormal embryonic sequestrations, which often go undetected for extended periods until triggering factors lead to sudden growth, resulting in pain or cosmetic deformity. Muscular trauma is thought to play a key role in the proliferation of pre-existing embryonic vascular malformations, which may help explain why most cases occur in younger populations. Localized vascular injury can result in the formation of granulation tissue, and when exacerbated by muscle activity, this proliferation may predispose to the development of a hemangioma. Hormonal changes have also been proposed as potential initiating factors [5]. A cosmetic deformity resulting from facial asymmetry is the most common reason for seeking treatment [18]. IMH usually present as slow-growing masses without symptoms or signs such as thrills, bruits, or pulsations, which would suggest their vascular origin. The surrounding muscular fibrosis may explain the absence of these signs and contribute to the mass's rubbery, firm textura [44]. Skin discoloration over the lesion is also uncommon. Pain at presentation is often associated with rapid growth and compression of nearby structures [45]. Clinically recognizable hemorrhage in IMH is considered rare. In 1997, Itosaka et al. reported a temporalis muscle hemangioma with a peculiar course of appearance and regression, which was believed to be caused by hemorrhage [15]. The differential diagnosis includes lipoma, neurofibroma, dermoid cyst, enlarged lymph nodes, soft tissue sarcoma, temporal arteritis, and myositis ossificans [19]. Nowadays, MRI is the most accurate noninvasive method for diagnosing IMH. It offers superior soft tissue contrast resolution compared to CT without contrast agents and allows for both sagittal and coronal imaging. IMH typically appear with low signal intensity, similar to muscle, on T1-weighted images, and high signal intensity, greater than fat, on T2-weighted images. Despite contrast enhancement, CT images may not always clearly differentiate hemangiomas from surrounding tissues. Ultrasound can be helpful for assessing superficial lesions, but both CT and ultrasound can detect the presence of phleboliths within IMH, which are calcified thrombi, a characteristic feature of this type of hemangioma [46].

Arteriography images do not offer adequate information regarding the relationship between hemangiomas and adjacent structures for precise surgical planning and are typically limited to high-flow lesions. However, arteriography remains important to evaluate the tumor's vascular supply and to facilitate preoperative embolization. Fine-needle aspiration is often inconclusive and may carry a risk of significant blood loss during the procedure, especially in cases of high-flow hemangiomas.Although the management of IMH should be individualized, surgical excision, including a rim of normal surrounding tissue, is the preferred first-line treatment according to several authors. This approach allows for histological evaluation of the mass and reduces the rate of local recurrence [19,20]. Other proposed treatment modalities include cryotherapy, steroid and sclerosing agent injections, arterial embolization, arterial ligation, and radiotherapy. Complete surgical resection is indicated in cases of tumor growth, pain, cosmetic deformity, thrombocytopenia, or suspicion of malignancy [47]. Heckl [20] and Uralogu [24] advocate for strict clinical and radiological follow-up for at least two years. Surgical intervention is typically reserved for capillary hemangiomas (due to their more aggressive behavior), progression of the lesion, or when quality of life is significantly affected by intractable pain, aesthetic concerns, or functional impairment[20]. Subselective arteriography is widely used for diagnosing and treating high-flow vascular malformations and hemangiomas. The complication rate associated with this technique has decreased, and severe adverse events such as stroke, cranial nerve ischemia, blindness, and significant hemodynamic changes have become less common. However, skin and mucosal necrosis remain recognized side effects of embolization [48-50].

Preoperative embolization through the lesion's arterial supply reduces blood flow within the angiomatous mass and minimizes blood loss during surgery, with a low complication rate. If surgery is performed immediately after embolization, proximal occlusion of blood vessels near the lesion can be done, as collateral blood supply is unlikely to develop within 72 hours [51] Embolization as a standalone treatment may have limitations, such as blood vessel recanalization and collateral vessel growth over time, which could allow for tumor regrowth and recurrence. Thus, a combined approach of embolization and surgical resection is recommended for IMH [48-50]. Local recurrence, primarily associated with incomplete resection, varies among the three pathological subtypes of IMH. Recurrence rates of

28%, 20%, and 9% have been reported for mixed-type, capillary, and cavernous IMH, respectively [14]. No one of our cases, nine and five years after treatment respectively, has shown any sign of recurrence.

Conclusion

Hemangiomas of the temporalis muscle are exceptionally rare, with only 37 well-documented cases in the international literature. Local recurrences have been reported, and are more frequent in the mixed and capillary types. Preoperative embolization and surgical excision including normal surrounding tissue seem to be the best choice of treatment for capillary and mixed hemangiomas, so that they reduce blood lost during surgery and local recurrences.

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